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Kanji-limited Pure Alexia

—A Case Report and the Problem
of Variability in Pure Alexia—

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SUMMARY

A case of Kanji-limited pure alexia has been reported. Concomitantly, we have examined another five pure alexic patients, administering the same linguistic test battery in order to compare the variability of disturbances in reading ability. It is concluded that the first case reported in detail may represent a pure example of categorization or global reading deficit. We suppose our six cases of pure alexic patients may be classified into three types as follows. Type I: Alexia is mild or moderate and in which letter-by-letter or analytical reading strategy might be predominant. Type II: Alexia is similarly mild or moderate but in which global reading strategy might be predominant. Type III: Degree of alexia is so severe that we could not judge the dominant reading strategy, or in other words, neither analytical nor global reading strategy could be used well.

INTRODUCTION

Pure alexia or alexia without agraphia is admitted generally as a clinical entity among various forms of alexia. However, whether pure alexia is from the neuro-linguistic point of view homogeneous or not has not yet been completely clarified. From the anatomo-clinical point of view, after Dejerine's and Geschwind's famous assertions (1892, 1965), Greenblatt (1976) has advanced typology of pure alexia. According to him there exist at least four variations of pure alexia: splenooccipital form with hemianopsia, splenooccipital form without hemianopsia, subangular form with hemianopsia, subangular form without hemianopsia. He insisted that all these forms could be explained by the disconnection between left angular cortex and bilateral visual primary cortex. This explanation may be plausible to the appearance or manifestation of reading disturbance itself, but various linguistic features which appear in pure alexic patients, for example, verbal alexia, literal alexia or very large varieties of Kanji-Kana reading impairments in Japanese pure alexic patients, cannot be explained only by the Greenblatt's anatomical disconnection

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theory.

Hécaen, Kremin and others (1976) have studied intensively this problem, and proposed that verbal alexia and literal alexia observed in pure alexia may probably reflect not the severity but the basic qualitative difference which exists in pure alexic symptom itself. This issue has not yet been examined sufficiently in Japanese patients, whose written language system is specific.

We have observed a Kanji-limited pure alexia. We investigated this issue in this patient and concomitantly examined another five pure alexic patients, administering the same linguistic test battery.

CASE REPORT

M.N. a 67 year-old right handed house-wife consulted us on October 26th 1979. She had been described as having had an atrial fibrillation from about the age of 60. The level of her reading capacity had been average before the onset of the disease; she had no difficulty in reading any article in a newspaper. In the early morning of October 25th 1979, she suddenly showed signs of confusional state with the short verbal utterance: "I feel strange...". On the initial neurological and neuropsychological examinations, no motor disturbance, sensory impairment, nor marked visual field defects by confrontation test were revealed. However, she presented a severe amnesic syndrome with marked disorientation for time and impairment of memory retention, a word finding difficulty with perseveration, incomplete right unilateral spatial neglect, picture agnosia, color agnosia, topographical disorientation and very noticeable global alexia without manifest agraphia.

With regard to the alexia, when presented with a Kana-word composed of four letters, for example ひこうき (Hi-Ko-U-Ki="airplane") written horizontally from left to right, she pointed out the two left-hand letters, i.e. Hi and Ko, and made the erroneous reading (Hi→To?, Ko→To?). If the examiner called her attention to the number of letters, she searched for and found the remaining two letters on the right-hand side. However, her reading of this word failed with perseverative paralexia. There was the same tendency in the reading of Kanji-words (for example, 時計 Tokei="watch", 新聞 Shinbun="newspaper"). These phenomena may reflect at least partly right unilateral spatial neglect, but she has finally succeeded in a cancellation test. She could write without hesitation, "山の上に大きな木があります"="There is a big tree on the mountain", on dictation, but approximately five minutes later *she could not read this same sentence which she had written herself just before*. In addition, this difficulty was not reduced by kinetic facilitation.

One week later, she was admitted to the Kyoto First Red Cross Hospital. We were very surprised by the fact that, in spite of her almost unchanged memory impairment and visual agnosia, her reading ability had become nearly normal for Kana (quasi-phonogram)-words but had not yet recovered for Kanji (quasi-

ideogram)-words. She could read without hesitation almost all Kana-parts of any sentence presented, but nevertheless stopped at Kanji-parts and hesitated to read them, with several paralexia. This difficulty in reading Kanji-letters was apt to increase for those which were composed of a greater number of written strokes.

CT scan were performed on October 27th and on December 5th 1979; both of these showed a localized low density area in the left occipito-temporal region with no indication in the medial side of the left occipital area (Fig. 1). This finding indicated a partial infraction (possibly due to embolism) of the posterior cerebral artery.

Ophthalmological examinations revealed very slight defects of the right upper parts of the visual field of the both eyes (Fig. 2). On the basis of the results of Farnsworth Panel D-25 Test, the existence of a tritanopic tendency was suspected (Fig. 3).

Further neuropsychological examinations

1) Reading capacity for Kanji-letters

a) A test of reading aloud 42 Kanji-letters, consisting of seven groups, composed respectively of Kanji-letters of 3, 5, 7, 9, 11, and 13 written strokes (=Test A), was administered to her. The results (Fig. 4) indicated that the more numerous the written strokes of Kanji-letters, the greater her reading difficulty became.

b) In spite of her inability to read these letters aloud, auditory-visual matching

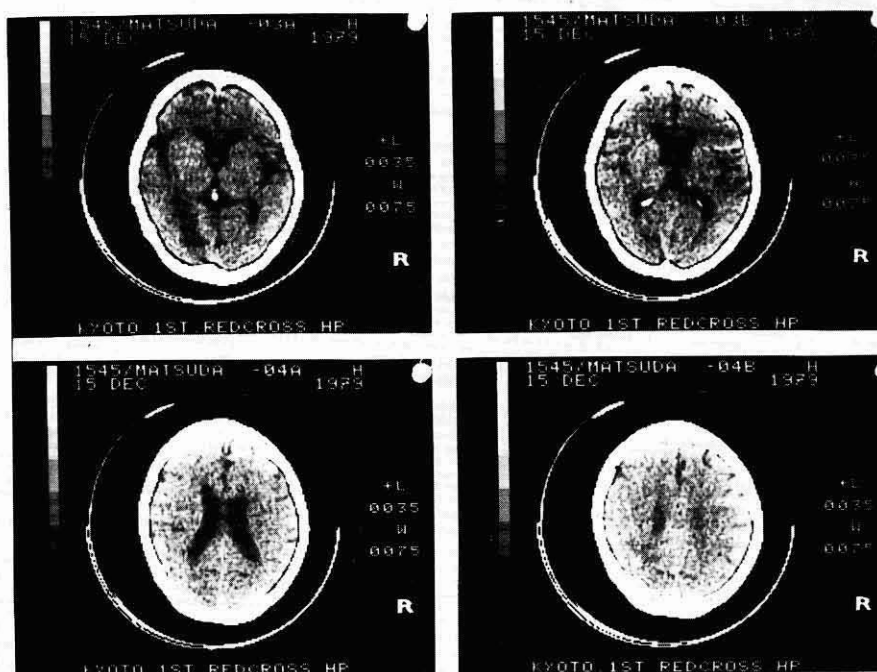


Fig. 1. CT scan finding (12-5, 1979).

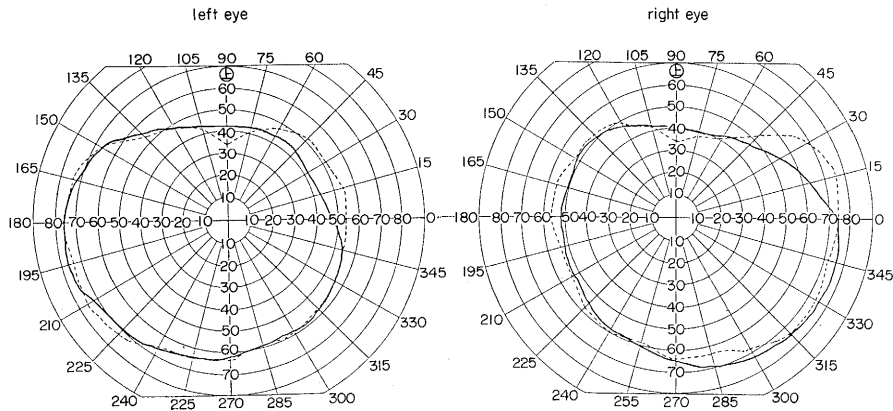


Fig. 2. Visual Field Examination (12-6, 1979)

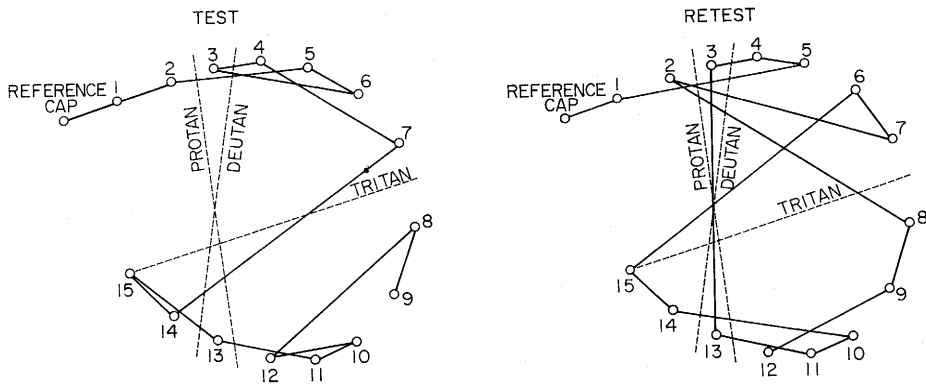


Fig. 3. Farnsworth Panel D-25 Test (11-5, 1979)

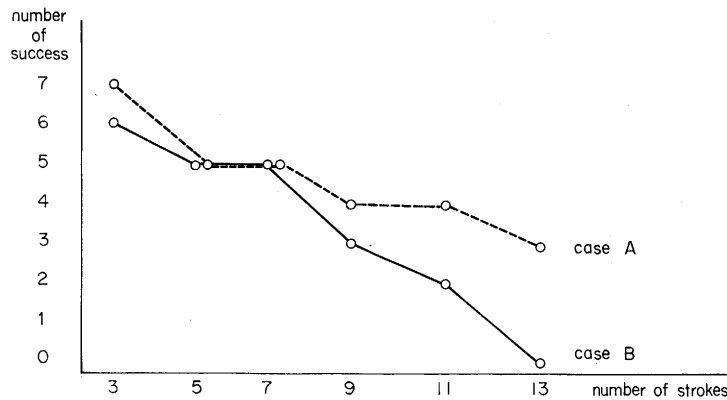


Fig. 4. Strokes-dependent Kanji-letter reading Test
(presented case=case A, and case B mentioned later)

of Kanji-letters among seven letters with the same number of written strokes was almost completely successful.

c) "odd word out test" (=Test B)

She should have pointed out the one odd Kanji-word in a set of six, five of which

belonged to the same category; one of these sets, for example, 歯 tooth, 眼 eye, 机 desk, 肩 shoulder, 鼻 nose and 耳 ear. In this set the odd word is naturally 机 desk, because all other words fall into the category of body parts. In ten normal subjects this test was found to be performed very easily. The patient showed no self confidence and could point out the correct odd word in only one set. However, if these six words were presented in succession as auditory stimuli, she made almost no mistakes.

2) Reading capacity for Kana-letters

She could read almost all Kana-words correctly and fluently, whether these were meaningful or meaningless. She cleared also with little effort the reading-aloud of ten meaningful and ten meaningless Katakana words composed respectively of two letters (=Test C).

3) Oral language

Her spontaneous speech was fluent without paraphasia and no impairment of repetition was found. In the situation of normal conversation, she could comprehend almost everything the examiner said. She executed Marie's Three Paper's Test successfully.

4) Writing capacity

In her spontaneous written manuscripts no significant errors occurred, except firstly a tendency to use relatively many Kana-letters instead of Kanji-letters, and secondly a few miscellaneous morphological errors in Kanji-letters.

Almost identical characteristics to those in her spontaneous writing were found in her dictations.

Summary of this case

During the first week the patient manifested global alexia without marked agraphia, but the alexia attenuated very rapidly and there remained only the Kanji-reading disability. Severe topographical disorientation, mild anomia, mild color agnosia and mild picture agnosia also continued.

RESULTS OBTAINED FROM ANOTHER FIVE PURE ALEXIC PATIENTS

We have examined concomitantly another five patients of pure alexia, administering the same test battery (Test A, Test B, Test C), in order to know the speciality of the case mentioned above and the possible variability of pure alexia.

Etiology of all cases were infraction of left posterior cerebral artery which were affirmed by CT scan. Age, sex, months after onset and educational level of each case (including the above presented case=case A) are indicated in Tab. 1.

Results of Test A (Strokes-dependent Kanji-letter reading test)

Case B presented almost the same pattern as that observed in case A (Fig. 4). On the contrary, the number of strokes of Kanji-letters did not seem to have any significant influence upon the results of reading aloud of Kanji-letters, in the case C and D (Fig. 5). There exists a same tendency in the case E and F, but the results

Table 1. Six cases of pure alexia after infarction of left PCA
(All cases are right-handed)

case	Age	sex	months after onset	education
A	67	f	1-3	middle school
B	68	m	1-3	high school
C	60	m	34-36	university
D	46	m	2-3	university
E	75	m	70-72	middle school
F	57	m	2-4	middle school

were worse in general than that in the case C and D (Fig. 6).

Results of Test B: (Kanji odd word out test).

The number of success and its percentage was relatively low in the case A and B, complete in the case C, D and E and moderate in case F (Tab. 2).

Results of Test C (meaningful and meaningless Katakana words reading test)

All items could be read finally in the case A, B and C and no items could be read in the case E and F. In the case D, eight out of ten meaningful but only one out of ten meaningless items could be read. Concerning the mean time for correct

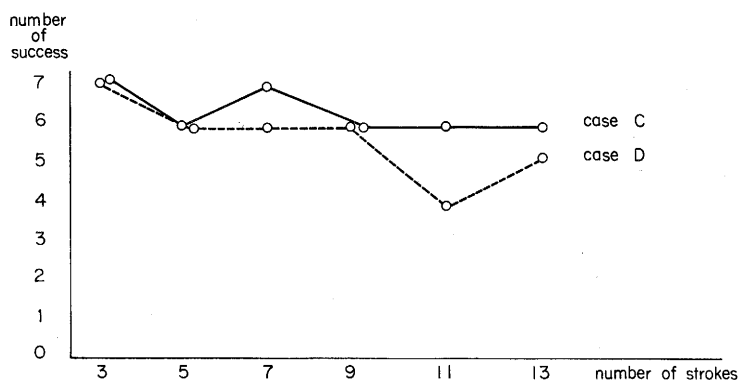


Fig. 5. Strokes-dependent Kanji-letter reading Test case C and D

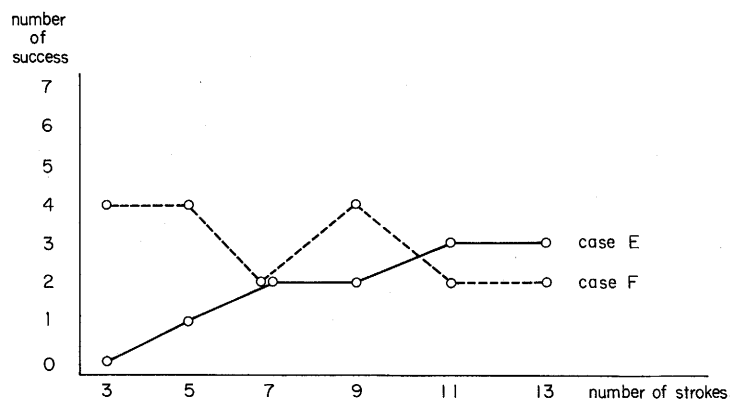


Fig. 6. Strokes-dependent Kanji-letter reading Test case E and F

Table 2. Results of Yanji Odd Word Out Test in the six pure alexic patients

case		%
A	1/6	16.7
B	2/6	33.3
C	6/6	100
D	6/6	100
E	6/6	100
F	4/6	66.7

Table 3. Meaningful and meaningless Katakana words reading test in the six pure alexic patients

case	meaningfull words	meaningless words	significance
A	10/10, 1.25 sec.	10/10, 1.34 sec.	NS
B	10/10, 4.45 sec.	10/10, 4.50 sec.	NS
C	10/10, 3.50 sec.	10/10, 8.70 sec.	S, $p < 0.001$
D	8/10, 6.80 sec.	1/10, 24.5 sec.	S, $p < 0.001$
E	0/10	0/10	/
F	0/10	0/10	/

reading, we found no significant difference between that for meaningful and meaningless items in case A and B. Oppositely, there exists the definite significant differences in the case C and D (Tab. 3).

DISCUSSION

There are several problems to be solved. 1) In the first place the case presented in detail is very exceptional because of rapid amelioration of alexia and of remained Kanji-limited alexia. How can explicate this rapid recovery? 2) Why did only the Kanji-alexia persist? 3) Is this case to be regarded simply as exceptional or this case suggested the existence of an "ideal type" of pure alexia? And lastly 4) Is the pure alexia a homogeneous neurolinguistic entity, or are there any subtypes of pure alexia, which are distinguished from each other?

Concerning the rapid recovery from alexia, Johansen et al. (1978) stated that in cases of lateral infraction of left posterior cerebrel artery the alexia is in general transient. Our case accorded with his view for the Kana-alexia. However, we must still explain the persistence of the Kanji-alexia. The disconnection hypothesis proposed by Geschwind, which postulates an interhemispheric interruption between the visual association cortex of the right hemisphere and the speech area of the left hemisphere, probably cannot give us a sufficient explanation for the manifestation of the alexia in our case, because the patient's right visual field was

fairly intact except for the very uppermost part. Nevertheless, the possibility that in the acute phase interhemispheric disconnection may have taken place by the mechanism of so-called diaschisis, cannot be denied. In any case another mechanism must be found to account for the occurrence of Kanji-limited alexia.

Does the persistence of Kanji-limited alexia in this case reflect the specificity of the Japanese written language system? Japanese is essentially syllabic and each Kana-letter generally represents one syllable. Therefore, the Kana-system does not precisely correspond to the alphabetic system of European languages, even though both systems have the common property of being composed of quasi-phonograms. On the other hand, there is nothing at all corresponding to the Kanji-system in European languages, because it has often been said that this system is in general ideographic. In addition several studies have already been published which have suggested the existence of differences between the ways Kana and Kanji are processed by normal subjects. These differences may be related to the phenomenon of Kanji-limited alexia in this case, but some proof of this is required.

Here, we have to consider the problem of the distinction or opposition between literal alexia and verbal alexia. This distinction was developed by the authors who used alphabetical language systems and although it has some relation to the problem of Kanji/Kana alexia in Japanese, this distinction is too simple to explain alexias in Japanese. One Kanji-letter almost always has literal and verbal value at the same time; one Kana-letter has a literal value in a different way from alphabetical letter.

Until about 1960, the distinction between literal alexia and verbal alexia in pure alexia had been considered as an expression of the severity of this symptom (Ajuriagurra et Hécaen, 1960), but recently this distinction has been reconsidered by Dubois-Charlier, Hécaen and Kremin (1972, 1974, 1976). Hécaen distinguished three qualitatively different types of pure alexia, i.e. literal alexia, verbal alexia and phrase alexia (1972). According to Kremin (1976), the fundamental disorder in literal alexia and phrase alexia is the impairment of "dénomination" or "séquentialisation", and that in verbal alexia is the impairment of "catégorisation". Hécaen and Kremin insist particularly on the different nature of literal alexia and verbal alexia.

Incidentally, it has often been said that there exist at least two different reading strategies (Alajouanine, 1960): global reading and analytical reading. One can immediately comprehend, for instance, one word or several words by means of global reading, but on the other hand analytical reading plays the role of, for example, distinguishing several morphologically similar words. In the case of the Japanese written language, it is very probably that if a sentence is written by Kana (Hirakana)-letters alone, analytical reading strategy will be more important than global reading. However, in a normally written sentence containing both Kana-letters and Kanji-letters one would principally use global reading strategy

to read Kanji-letter parts, because each Kanji-letters has in general one or more semantic values and these values could immediately be visually grasped as a whole without great need for the use of analytical reading processes.

It seems to us that reading by visual categorization processes and visual sequentialization processes as stressed particularly by Kremin and Hécaen might respectively bear a strong resemblance to global reading strategy and analytical reading strategy mentioned above.

Our first patient showed a fairly bad score on the "odd word out test" executed visually, but in contrast a very good score when the test was executed by auditory means. This fact may suggest that her capacity for visual categorization in reading was deficient. Secondly, the fact that on the strokes-dependent Kanji-letter reading test she presented the greater difficulty the more numerous the written strokes of Kanji-letters became, suggests also the deficiency of her lower capacity of visuo-linguistic categorization, because it seems that the more numerous written strokes of Kanji-letters the more predominant the strategy of global reading might become. Thirdly, that she could read Katakana-words almost in the same way no matter which is meaningful or meaningless, seems to show the absence of important role of global reading strategy in her reading processes, because if the global reading strategy exists not negligibly the meaningful words must be read easier than the meaningless words.

Since visual categorization deficit might be taken as similar to global reading deficit, it could be presumed that the Kanji-limited alexia in this case might be related to global reading deficit with almost intact analytical or sequentialization reading process. That is to say, this case may represent a pure example of categorization or global reading deficit.

Although almost all case reports of pure alexia in Japan say that patients manifested both Kanji and Kana reading disturbances in varying degrees (Torii, 1979 et al.), the very existence of exceptional cases such as our patient suggest strongly that pure alexia has some neurolinguistic miscellaneous subtypes.

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- A. mild or moderate alexia
 - type I (case A and case B)
 - letter-by-letter reading dominant form
 - Kana-reading > Kanji-reading
 - type II (case C and case D)
 - global reading dominant form
 - Kanji-reading > Kana-reading
 - B. severe alexia
 - type III (case E and F)
 - miscellaneously mixed form
 - Kanji-reading > Kana-reading
-

Fig. 7. Classification of pure alexia

According to these arguments and results obtained from another five cases, we tried to classify our six cases into three types as follows (Fig. 7); case A and B may belong to type I, in which alexia is mild or moderate and letter-by-letter or analytical reading strategy might be used predominantly, because in these cases there exists no significant differences between meaningful and meaningless Katakana-words reading ability, and global reading strategy might be poorly used because of bad scores of "odd word out test" and of negative influences of number of strokes by Kanji-letter reading.

In contrast to type I, case C and D may belong to type II, in which alexia is similarly mild or moderate, but global reading strategy might be predominantly used, and reversely letter-by-letter reading strategy be used in lesser degree, because of the presence of significant differences of reading ability between meaningful and meaningless Katakana-words, of good score of "odd word out test" and of apparent opposite tendency on the strokes-dependent Kanji-letter reading test, to that in case A and B.

In case E and F, degree of alexia is so severe that we could not judge the predominant reading strategy used by patients. These cases are named type III, in which reading strategy may variedly impaired.

We know that in the past published cases of pure alexia in Japan the dissociation of Kanji-Kana impairment is very much varied as mentioned above. These phenomenon could be explained by such hypothesis of neurolinguistic heterogeneity of pure alexia.

We proposed a psycholinguistic model of reading process (Fig. 8) which we modified that proposed by Newcomb et al. (1975). We have added visuo-linguistic process, composed of visual global reading process and of visual analytical reading

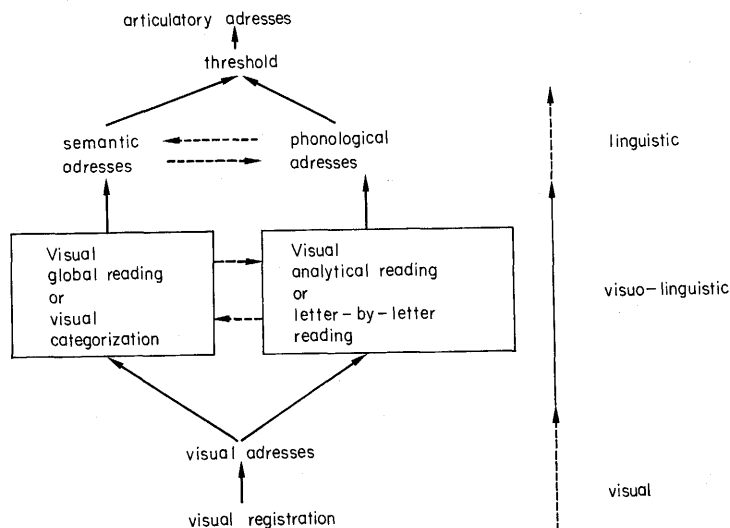
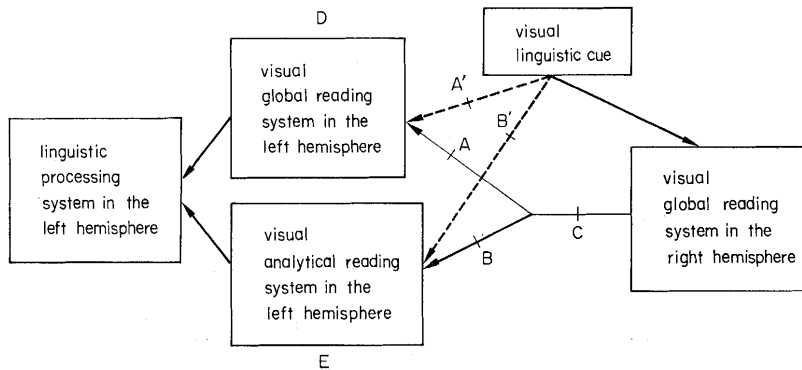


Fig. 8. A psycholinguistic model of reading process



A or A', D: pure alexia type I
 B or B', E: pure alexia type II
 A + B, A' + B', C, D + E: pure alexia type III

Fig. 9. A neurolinguistic model of pure alexia

process just before the proper linguistic reading process. We believe this added stage might be essential for the appearance of heterogeneity of pure alexia.

Now we propose a neurolinguistic model of pure alexia. If patient has right hemianopsia, visual linguistic cue addresses at first only into the right hemisphere. Some split-brain studies suggest that right hemisphere may have very limited reading comprehension ability which should be amorphous or roughly global. Here, we presume that in the left hemisphere exist some visual global reading system and some visual analytical reading system. In a case without hemianopsia visual linguistic cue might reach directly these two systems. Referring to the Fig. 9 if the pathway A or A' is interrupted, or system D proper is damaged and another portions are almost preserved or intact, reading strategy may be limited to global reading comprehensions by right hemisphere and visual analytical reading by left hemisphere, and consequently patient's dominant reading strategy may become analytical or letter-by letter. On the contrary, the pathway B or B', or system E proper is damaged, and another portions are almost preserved or intact, visual global reading by right and left hemisphere become predominant. In the former circumstance pure alexia type I might appear and in the latter type II appear.

If the pathway C, or A and B, or system D and E proper are impaired significantly, alexia might become very severe and according to the severity of impairment and to the site of lesion, alexia might take various forms. This situation correspond perhaps to the pure alexia type III.

Recently, Uchiyama et al. (1987) denied the existence of any subtypes of pure alexia from the viewpoint of reading strategy. But we believe that it is necessary to regard the variability of reading strategy for such an "ideal exceptional case" as the case M.N. reported above in detail.

In any case, this hypothesis is certainly not yet confirmed, since whether this neurolinguistic model and neuroanatomical findings are compatible or not has not

yet made sure at present unfortunately.

Therefore we must continue our study in order to affirm or refine this neuro-linguistic model about heterogeneity of pure alexia.

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